

CASE REPORT

Choreoathetosis after subarachnoid hemorrhage related to an aneurysm of the posterior fossa

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INTRODUCTION

Non-traumatic subarachnoid hemorrhage (NSAH) is a neurological emergency with a high rate of death and complications.^{1,2} A ruptured intracranial aneurysm accounts for approximately 80% of NSAH cases.

The classic clinical manifestations of NSAH are headache, nausea and vomiting, focal neurological signs, meningeal irritation, and a reduction in the level of consciousness. In the present work, we describe a case of choreoathetosis that developed in a young patient who presented with a subarachnoid hemorrhage (SAH) related to an aneurysm of the posterior fossa. We also review the corresponding literature.

CASE REPORT

A 17-year-old boy, with no previous co-morbidity or neurological disability, reported the sudden onset of a severe headache associated with an alteration of his level of consciousness and meningeal signs. Upon admission to our department, three days after the acute event, he was confused, dysarthric, exhibited choreoathetoid movements of the distal upper limbs, exhibited postural instability, and exhibited the inability to walk, stand up, or sit up without help. The choreoathetosis began just after the onset of the SAH. The results of the motor, sensitivity, and reflex examinations were normal. The patient's hyperkinesia ceased with sleep. In addition, there was no family history of movement disorders.

A computed tomography (CT) scan revealed a hemorrhage in the fourth ventricle but no evidence of a parenchyma lesion. A cerebral angiography (Figure 1A) revealed a small saccular aneurysm of the basilar artery, which was located close to the emergence of the anterior inferior cerebellar artery (AICA). No evidence of vasospasm was observed.

On the fourth day after the ictus, the patient was administered haloperidol, and a progressive reduction in the frequency of choreoathetoid movements was observed. However, the dysarthria and postural instability remained. The patient was administered phenytoin for seven days

prior to the endovascular aneurysm repair, which was completed uneventfully. A control CT scan did not reveal hydrocephalus.

At the one-year follow-up visit, the patient did not exhibit any choreoathetoid movements; however, the cerebellar alterations remained, with an important static and dynamic imbalance and significant dysmetria. Magnetic Resonance Imaging (MRI) at the one year follow-up revealed cerebellar atrophy (Figure 1B).

DISCUSSION

Stroke-related movement disorders are uncommon (3.6%) and are very rare in SAH cases. Chorea, tremor, dystonia, Parkinsonism, and myoclonus have all been associated with cerebral infarcts and hemorrhaging.³ Movement disorders, which represent a portion of the clinical spectrum of acute stroke, may be delayed or progressive.

The first case of a movement disorder (chorea) after a SAH was reported by Sakai et al.⁴ and occurred eight days after the SAH onset. The CT scans revealed a SAH with ventricular dilation and periventricular lucency involving the bilateral caudate nuclei. The chorea was attributed to the vasospasm and hydrocephalus.⁴

In another case, reported by Morigaki et al.,⁵ the involuntary movements began shortly after the SAH onset. There was no acute hydrocephalus, and the authors attributed the symptoms to a hematoma on the corpus callosum that resulted from a rupture of an aneurysm of the distal accessory anterior cerebral artery. The hyperkinetic involuntary movements were suggested to have occurred due to the interruption of the cortico-striato-pallido-thalamo-cortical feedback loop.

Alarcón et al. analyzed 1,500 consecutive stroke patients over a period of ten years to identify patients with a movement disorder, which was observed in only 56 patients (3.6%). Chorea was the most common movement disorder (35.7%). Thirty-nine (69.6%) patients experienced an ischemic stroke, 14 (25%) experienced a parenchymal hemorrhage, and only three (5.3%) experienced a SAH. All of the patients with a movement disorder that was secondary to a SAH in the Alarcón et al. series presented with a tremor (Table 1) as their principal involuntary movement.³

The time that elapses between a stroke onset and the development of a movement disorder is variable. Alarcón et al. observed involuntary movements that began on the first day of the stroke in 12.5% of their patients but reported

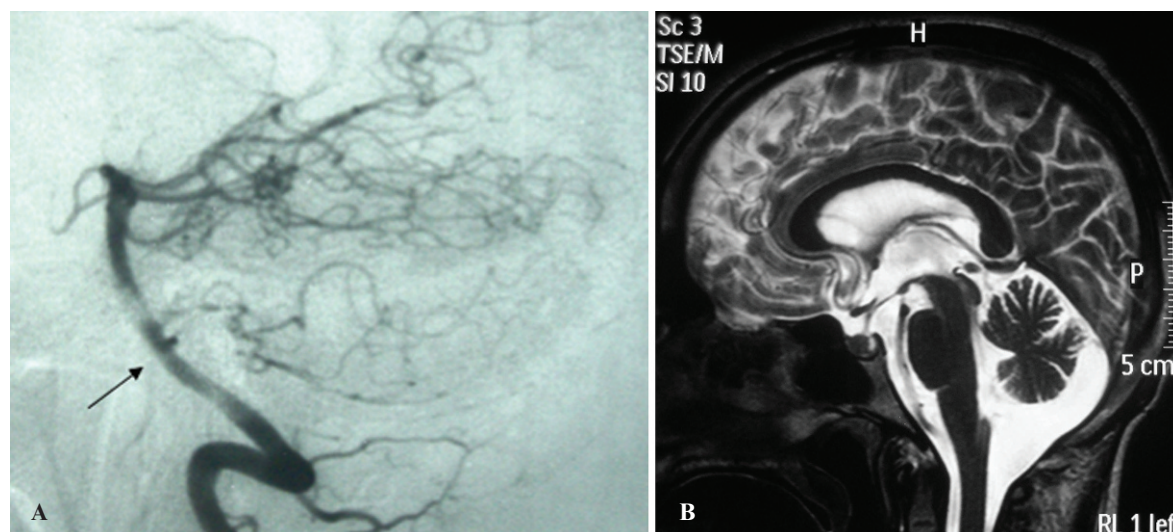


Figure 1 - A) Cerebral angiography at admission revealed a small saccular aneurysm of the basilar artery near the emergence of the anterior inferior cerebellar artery (AICA). B) T2-weighted Magnetic Resonance Imaging (MRI) at the one-year follow-up revealed cerebellar atrophy.

Table 1 - Movement disorders after SAH.

| Author (Year) | Age | Sex | MD | Time from SAH to MD | Aneurysm localization | MD Recovery | Hypothesis |
|-------------------------------------|-----|-----|--|---------------------|----------------------------------|-------------|--|
| Sakai et al. (1991) ⁴ | 71 | F | Chorea | 8 days | ICA - AChA | Total | Vasospasm Hydrocephalus |
| Alarcón et al. (2004) ³ | 74 | F | Tremor | * | ** | ** | Intraventricular hemorrhage |
| Alarcón et al. (2004) ³ | 55 | F | Tremor | * | ** | ** | Hydrocephalus |
| Alarcón et al. (2004) ³ | 63 | F | Tremor, dystonia, ataxia and dysmetria | * | ** | ** | Hydrocephalus |
| Morigaki et al. (2008) ⁵ | 72 | F | Choreoathetosis | Ictus | Distal accessory ACA | Total | Corpus callosum hematoma |
| Pereira et al. (2011) | 17 | M | Choreoathetosis | Ictus | Basilar artery at AICA emergence | Total | Disturbance of the dentato-rubro-thalamo-cortical pathways |

F = female; M = male; MD = movement disorder; AICA = anterior inferior cerebellar artery; ICA - AChA = internal carotid artery - anterior choroidal artery; ACA = anterior cerebral artery.

*Only time in days (18.7 days, SD = 12.8 days) reported between the diagnosis of stroke and the onset of abnormal movements in a group of 14 patients with tremor after stroke. Six patients experienced an ischemic stroke, five had parenchymal hemorrhage, and three experienced a subarachnoid hemorrhage.

**not mentioned.

cases in which the abnormal movement occurred much later, including Parkinsonism that began ten months after the stroke.³

While analyzing the literature data on movement disorders after a SAH (Table 1), we observed a total of six patients (including this reported case). Of these six patients, five were female and one was male, with a mean age of 58.6 years (SD 21.6, ranging from 17 to 74 years). Chorea or choreoathetosis was observed in three of these cases; tremor was observed in the other three cases. One patient in the tremor group exhibited associated dystonia. Our case is very atypical because he is the youngest patient ever described and was the only male to present with SAH-related involuntary movement.

In our case, no vasospasm, hydrocephalus, or even direct injury to the basal ganglia was observed that could explain the choreoathetosis. We suggest that the hematoma that was

located in the fourth ventricle may have disturbed the dentato-rubro-thalamo-cortical pathways, leading to transient choreoathetoid movements. Another hypothesis, without evidence of a vasospasm, is that the movement disorder was secondary to an ischemic lesion in the thalamogeniculate artery (a branch of the posterior cerebral artery), which disturbed the posterior ventral thalamic areas related to the basal ganglia circuit.

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